


CASE REPORT

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Treatment approach with clinical follow-up in monostotic fibrous dysplasia: a case series

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Abstract

Background Long bones are one of the most common sites involved in fibrous dysplasia. In some cases, there is no deformity, but patients suffer sustained or intermittent dull pain.

Case presentation Since 2021, a retrospective case series of seven East Asian patients with fibrous dysplasia of a long bone without severe deformity were reviewed. These patients include three male and four female patients, with an average age of 32.7 years (range 4–70 years). Fibrous dysplasia was diagnosed at a total of three different sites in these seven cases including five femurs (71.4%), one humerus (14.3%), and one fibular bone (14.3%). All patients received treatment with allogenic bone grafting or cortical strut grafting with or without compression locking and screw fixation. The radiological and clinicopathologic presentation was analyzed by the surgeon. There were no cases with polyostotic forms or fibrous dysplasia in combination with extraskeletal disease. The presenting complaint was pain in all cases and localized swelling in 1 (14.3%) of the cases.

Conclusions Autogenous fibular cortical strut grafting and compression hip screw fixation achieved good postoperative function and provided an early return to work for adult patients with fibrous dysplasia of the femoral neck with mild but prolonged symptoms. However, total hip replacement may be a suitable method for fibrous dysplasia of the femoral neck accompanied by pathological fracture.

Keywords Fibrous dysplasia, Radiological presentation, Clinicopathologic presentation, Allogenic bone grafting, Surgical treatment, Pathological fracture

Background

Fibrous dysplasia is a rare benign intramedullary fibro-osseous lesion with characteristics of fibro-osseous proliferation with intervening areas of normal or immature bone [1]. In these lesions, normal lamellar bone is replaced with regular trabeculae of woven bone accompanied by mature collagenous tissue, which often leads to substantial loss of mechanical strength

[2]. Subsequently, patient experiences pain, deformity, and even pathological fractures. Actually, fibrous dysplasia may affect one bone or multiple bones, and may occur in isolation or in combination with extraskeletal disease [3]. It is confirmed that postzygotic gain-of-function mutations in the *GNAS* gene cause persistent stimulation of adenylcyclase and dysregulated production of cyclic AMP and downstream signaling [4]. Although malignant transformation of fibrous dysplasia is rare, clinical management in fibrous dysplasia is also challenging. Besides, multiple barriers exist to providing early diagnosis and adequate treatment. The previous literature reported the clinical results of surgical treatment for fibrous dysplasia of the bone, including allogenic bone grafting or autogenous cortical strut grafting with or without compression locking plate and

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screws [5–7]. However, there are no therapeutic consensus about isolated monostotic bone lesions. In our cases, clinicopathologic outcomes were analyzed radiographically in populations with fibrous dysplasia of a long bone who accepted a treatment of allogenic bone grafting or cortical strut grafting with or without compression locking plate and screw fixation.

Methodology

Our study was approved by the institutional review board at our institution, and the patients permitted the use of their medical information. All patients provided informed consent according to ethical standards. We conducted a retrospective case series review from 2021 to 2023. Within the mentioned period, there were seven cases diagnosed as fibrous dysplasia on the basis of plain radiographs, computed tomography (CT), magnetic resonance imaging (MRI), and pathological findings.

The surgical procedure used in the present cohort is as follows. The patient is positioned on a surgical table, allowing the C-arm fluoro-image to be positioned between surgical lesions. A suitable straight incision is made over the long bone with diseased lesions. After communication with the patient, curettage and allograft, with or without fixation, were performed. At the beginning of the bone grafting, tissues of the tumorous

lesion are obtained and subjected to examination of the frozen section.

Case presentation

Patient 1

A 49-year-old East Asian woman complained of persistent dull pain in her left thigh for 6 months, which worsened during activity and disrupted her normal life. She described left lower limb fatigue, without numbness or motion disorder. She had no clinical history of chronic lumbar pain and traumatic history. A neurological examination and physical inspection were unremarkable. The plain radiographs, CT, and MRI suggested fibrous dysplasia in her left femur bone (Fig. 1). The histological examination confirmed the diagnosis of fibrous dysplasia of her left proximal femur bone (Fig. 2A). Then, curettage and bone allograft were used as an individual therapeutic approach (Fig. 3A). Time of initiation for full weight-bearing walking was determined according to the patient's symptoms and the radiographic findings on follow-up.

Patient 2

A 35-year-old East Asian woman complained of intermittent pain in her left thigh for 1 year. Her pain could not be relieved by means of bed rest, but it did not worsen during activity; however, it disrupted her normal life. No clinical history of related fibrous dysplasia such as chronic pain. In her physical examination, the physician observed

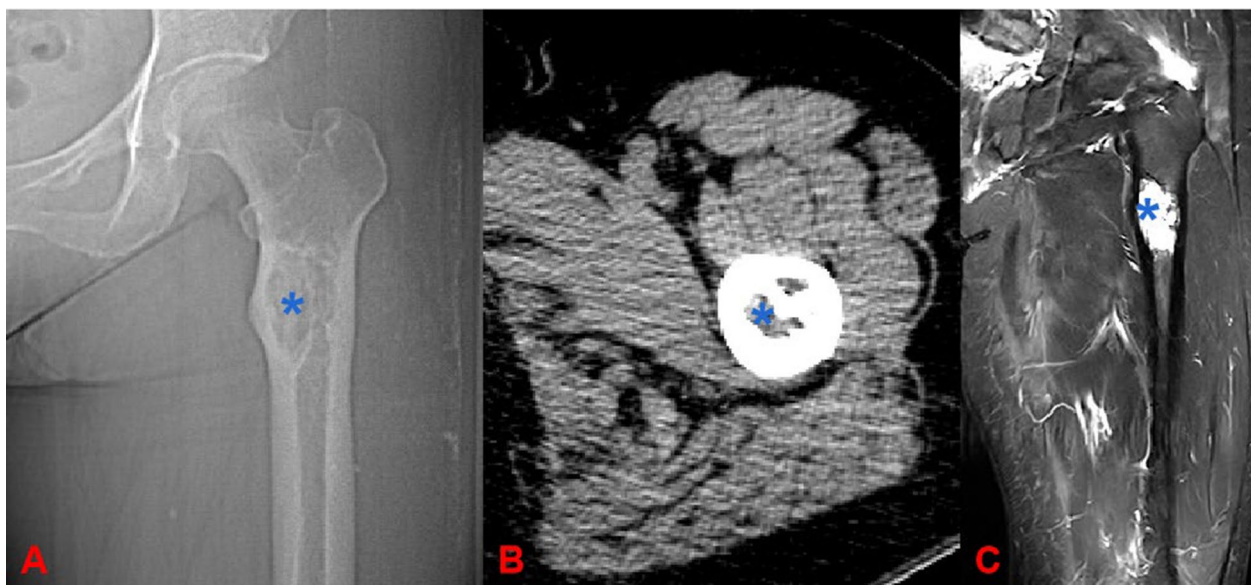


Fig. 1 The radiographic results of Patient 1. **A** The preoperative anteroposterior X-ray showed a ground-glass appearance (blue asterisk) of the left proximal femur bone. **B** The preoperative transverse computed tomography image showed the appearance of a lesion site in the marrow cavity of the proximal femur bone (blue asterisk). **C** The preoperative transverse T2-weighted (magnetic resonance imaging) image showed high intensity in the medullary space of the left proximal femur bone (blue asterisk)

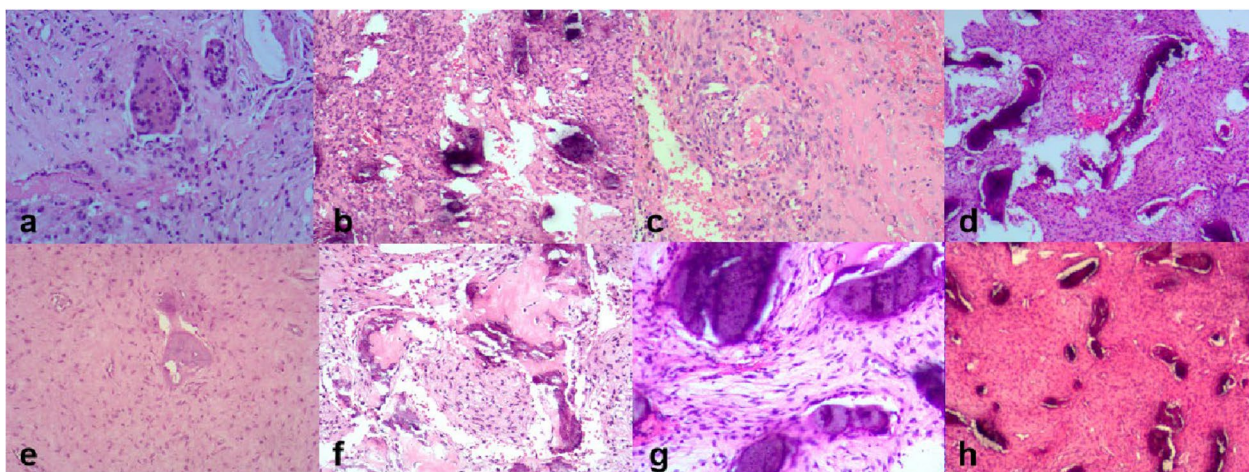


Fig. 2 The result of histological examination of our seven cases. **A–H** Hematoxylin and eosin staining, magnification 40x. All pathological examinations of our cases showed the typical appearance of fibrous dysplasia. The lesion contained some irregular immature bone trabeculae scattered throughout fibrous tissue. **B** and **D** Fibrous dysplasia accompanied by bone cyst. Solid areas and cystic spaces filled with blood, with cellular septa, and hyperplastic fibrous cells. **G** and **H** The first result (**F**) of fibular fibrous dysplasia and final result of recurrent fibular fibrous dysplasia showed the same appearance of irregular trabecular bone and hyperplastic fibrous tissue with a woven bone formation

tenderness and percussion pain of the left hip joint. The results of the neurological examination were unremarkable. The X-ray, CT, and MRI suggested multiple fibrous dysplasia in her left femur bone (Fig. 5). Fibrous dysplasia accompanied by bone cyst was also confirmed through pathological examination (Fig. 2B). The curettage and bone allograft along with internal fixation were performed in her left femur (Fig. 3B). Regular outpatient medical consultation was executed for her follow-up duration. According to result of X-rays, after 1 year, her internal fixation was removed by the surgeon (Fig. 4A).

Patient 3

A 37-year-old man complained of persistently severe pain in his right hip for 1 week, which limited his activity of daily living. He had no traumatic history and tumor-related history. In his physical inspection, the physician observed serious tenderness, percussion pain, and obstructed right hip joint activity. The radiographic images suggested fibrous dysplasia in his right femoral neck (Fig. 6). This diagnosis was also confirmed by pathological examination (Fig. 2C). The total hip replacement was performed in his right hip joint (Fig. 3C). A 1-month telephone follow-up proved that his condition was stable.

Patient 4

A 16-year-old East Asian female patient was admitted due to persistent pain in the right hip and abnormal gait over the previous 2 months. She had no history of present or past illness. Physical inspection indicated that the right hip had pain upon pressing and tenderness. The

radiographic images showed a ground-glass appearance with cortical scalloping and expansion of the right proximal femur and femoral neck (Fig. 7). Pathological examination revealed fibrous dysplasia along with aneurysmal bone cysts in her right proximal femur bone (Fig. 2D). The therapeutic method used involved curettage and bone allograft along with fixation of compression screws (Fig. 3D). Follow-up at 15 months proved that her condition was stable. Then, removal of internal fixation was performed by a surgeon (Fig. 4B).

Patient 5

A 71-year-old East Asian woman was observed to have proximal humeral osteolysis on outpatient service. She had a history of transient dizziness and persistent shoulder pain. Physical examination indicated that the right hip had pain upon pressing and tenderness. The radiographic images suggested fibrous dysplasia in her left proximal humeral bone (Fig. 8). Pathological examination confirmed fibrous dysplasia of the left proximal humeral bone (Fig. 2E). The same surgical technique was used for the humeral lesion site (Fig. 3E). Follow-up of 1 year proved that her condition was stable. After that, removal of internal fixation was performed by a surgeon (Fig. 4C).

Patient 6

A 17-year-old East Asian adolescent male patient complained of slightly intermittent pain in his right femur for 3 years that did not disrupt his normal life. He had no clinical history of past or present illness and had no traumatic history. A neurological examination and



Fig. 3 The therapeutic strategies fibrous dysplasia in our cases. **A** and **F** The postoperative anteroposterior X-rays showed the radiographically apparent refilling of bone grafts in ground-glass and partial radiolucent sites. **B**, **D**, and **E** The postoperative anteroposterior X-rays showed curettage and bone allograft along with internal fixation in these lesion sites. **C** The postoperative anteroposterior X-rays showed the total hip replacement had been performed in her right hip joint

physical inspection were unremarkable. The radiographic images suggested fibrous dysplasia in his right femur bone (Fig. 9). Pathological examination confirmed the diagnosis of fibrous dysplasia (Fig. 2F). Only the curettage and bone allograft were used in

this lesion site without internal fixation (Fig. 3F). Telephone follow-up of 3 months proved that his condition was stable.

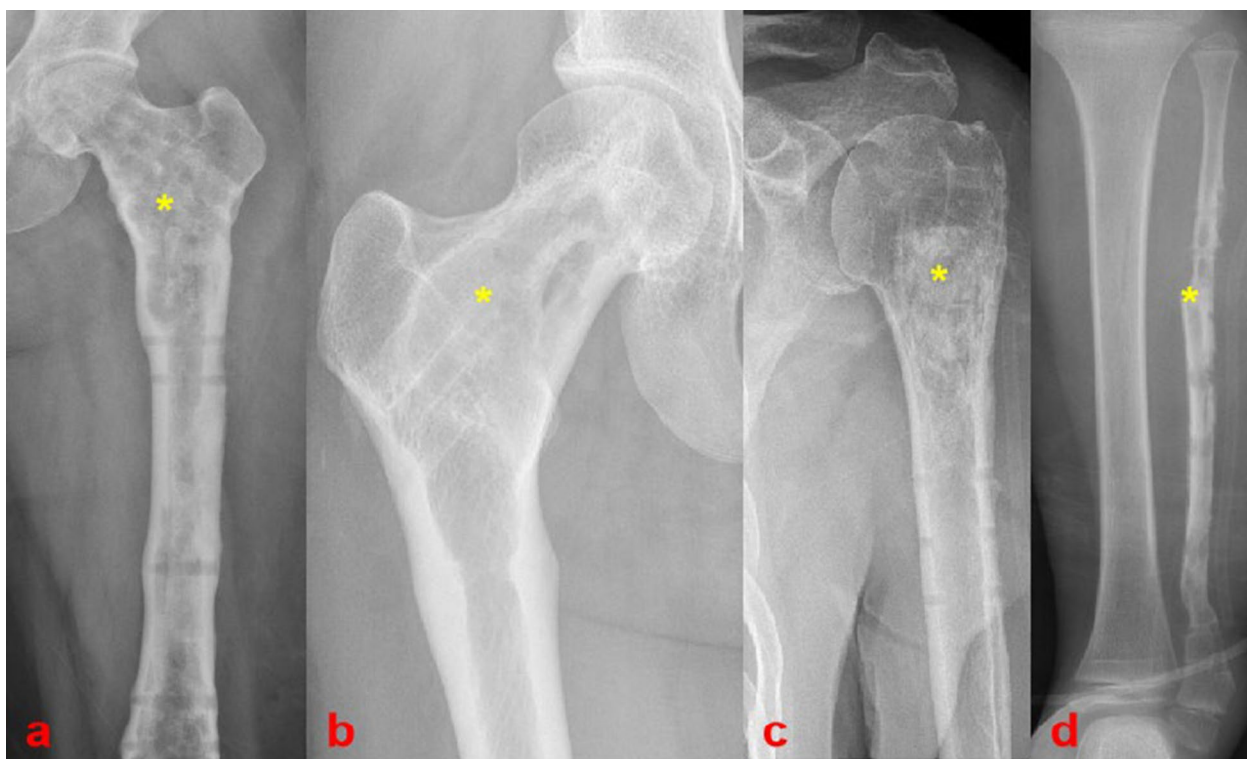


Fig. 4 The postoperative X-rays of our cases with removal of internal fixation. **A** The postoperative anteroposterior X-ray showed bone fusion (yellow asterisk) in a previous lesion site with a radiographically ground-glass appearance for Patient 2. **B** and **C** The postoperative anteroposterior X-ray showed removal of internal fixations and bone fusion (yellow asterisk) in previous lesion sites with a radiographically ground-glass appearance for Patient 4 and Patient 5, respectively. **D** For Patient 7, a strong autogenous fibular bone fused to allogenic fibular bone (yellow asterisk) was observed, and the internal fixation was removed by the surgeon

Patient 7

A 4-year-old East Asian boy was admitted for persistent pain in the left lower limb and abnormal gait over the previous 9 months. He had no history of present or past illness. The X-ray images suggested fibrous dysplasia in his left fibular bone (Fig. 10A). After the rapid intraoperative pathological examination suggested fibular fibrous dysplasia (Fig. 2G), removal of tumor tissue in the fibular bone marrow cavity was performed by means of curettage (Fig. 10B). Then, a bone allograft was implanted into the fibular medullary cavity (Fig. 10B). Nevertheless, he was readmitted with similar clinical symptoms including persistent pain, abnormal gait, and local swelling at the age of 6 years old. He was diagnosed with recurrent fibular fibrous dysplasia on the basis of the second medical examination (Figs. 2H, 10C). He underwent fibular bone tumor radical resection and fibularis longus allograft transplantation combined with locking plate and screws in his fibula bone because of recurrent fibular fibrous dysplasia (Fig. 10D). The length of the strut fibula graft was 13.9 cm. Good host bone for allogenic bone graft fusion was observed by the physician on postoperative regular follow-up (Fig. 10F). Finally, internal fixation was

removed by a surgeon when the patient was 8 years old (Fig. 4D).

Results

Within the mentioned period, there were seven cases diagnosed as fibrous dysplasia (Table 1). There were cases in three male (42.9%), and four female (57.1%) patients with an average age of 32.2 years (range 8–70 years). Fibrous dysplasia was diagnosed at a total of three different sites in these seven cases including five in the femur (71.4%), one in the humerus (14.3%), and one in the fibular bone (14.3%) (Table 2). There were no cases with polyostotic types or fibrous dysplasia in combination with extraskeletal disease. The presenting complaint was pain in all cases and localized swelling in one (14.3%) of the cases. Clinically pathological bone fractures were encountered in one case (Patient 3). Five of these cases were localized to the femur (Patients 1, 2, 3, 4, and 6), and one case showed multiple location of lesions of the femur bone (Patient 2).

One case was diagnosed as fibrous dysplasia and underwent curettage of the lytic lesion localized in the patient's left fibular bone alone (Patient 7). Although, the

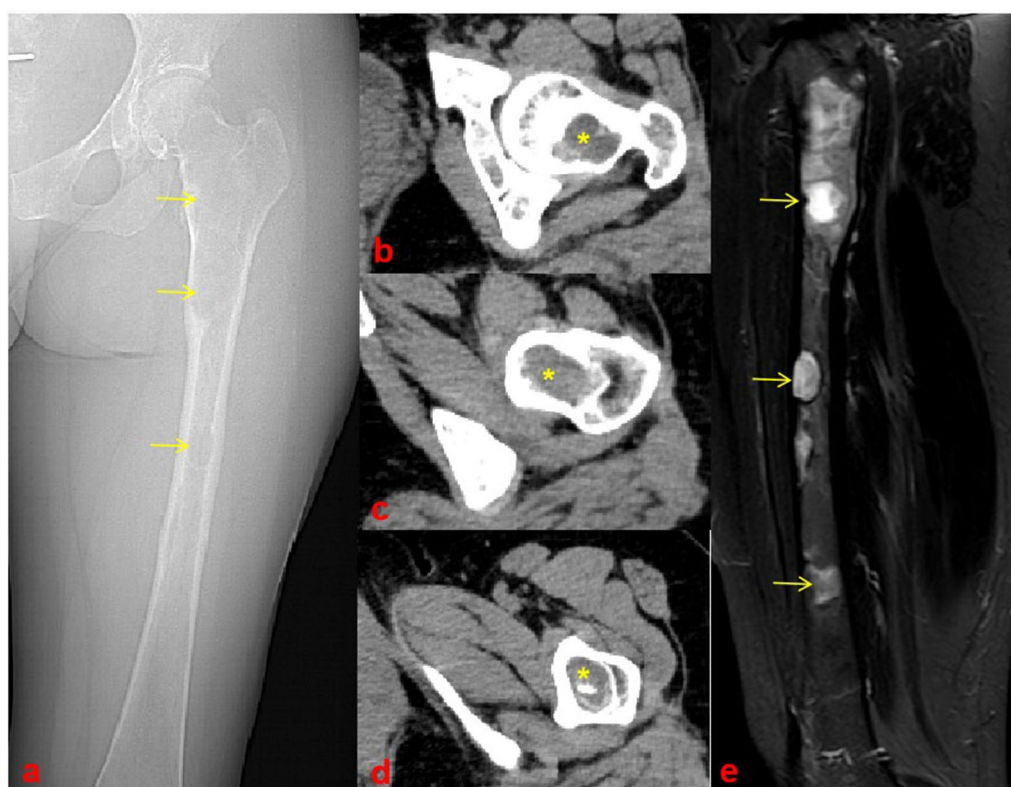


Fig. 5 Fibrous dysplasia accompanied with bone cyst in Patient 2. **A** Preoperative radiographic image showed multiple lesion sites with a ground-glass appearance (yellow arrows) in the femur bone for Patient 2. **B–D** Preoperative axial computed tomography images showed the appearance of lesion sites in the marrow cavity of the proximal femur bone (yellow asterisks). **E** Preoperative magnetic resonance imaging. The preoperative sagittal T2-weighted image showed multiple high intensities in the medullary space of the femur bone (yellow arrows)

previous pathological examination revealed benign features, the subsequent detailed radiological examination revealed recurrent monostotic fibular fibrous dysplasia during follow-up. Then, this child underwent radical surgery and fibularis longus allograft combined with fibular locking plate and screws. Two cases were diagnosed as fibrous dysplasia accompanied with bone cyst; one of the cases was aneurysmal bone cysts localized in the right proximal femur (Patient 4). One case underwent total hip replacement attributed to pathological fracture of the femoral neck (Patient 2).

We obtained the radiological images of all cases from the archive. Histopathologically, seven cases were classical fibrous dysplasia. Microscopically, they showed typical irregular woven bone trabeculae with irregular, anastomosing immature bone trabeculae scattered throughout a fibrous tissue without malignant features (Fig. 2). An 8-year-old patient had a fibular lesion localized at the left fibular bone showing recurrent monostotic fibular fibrous dysplasia (Patient 7).

The first line of treatment consisted of curettage and bone grafting with or without internal fixation. In our cases, two patients accepted curettage and bone grafting

without internal fixation (Patients 1 and 3), and four patients receive curettage and bone grafting with internal fixation (Patients 2, 4, 5, and 7). One case underwent total hip replacement (Patient 3). One patient was diagnosed with fibrous dysplasia associated with bone cyst accompanied with pathological fractures of the femoral neck (Patient 4). In one case, the patient underwent radical resection because of recurrent fibrous dysplasia during follow-up (Patient 7). No problems were encountered on follow-up in these seven cases. Mean follow-up after surgical treatment was 13.85 months (range 1–39).

Discussion

As a rare benign skeletal disorder, fibrous dysplasia presents with a unique non-hereditary and idiopathic nature, and shows histomorphologic characteristics of normal bone replacement by fibro-osseous tissue [1]. Actually, it can lead to decreasing exercise capability and a consequently high susceptibility to fractures and breaks in some acute cases. In fact, some cases with fibrous dysplasia are often discovered by a physician on routine physical examination or other medical practice because of its occult clinical manifestation. The accurate incidence of

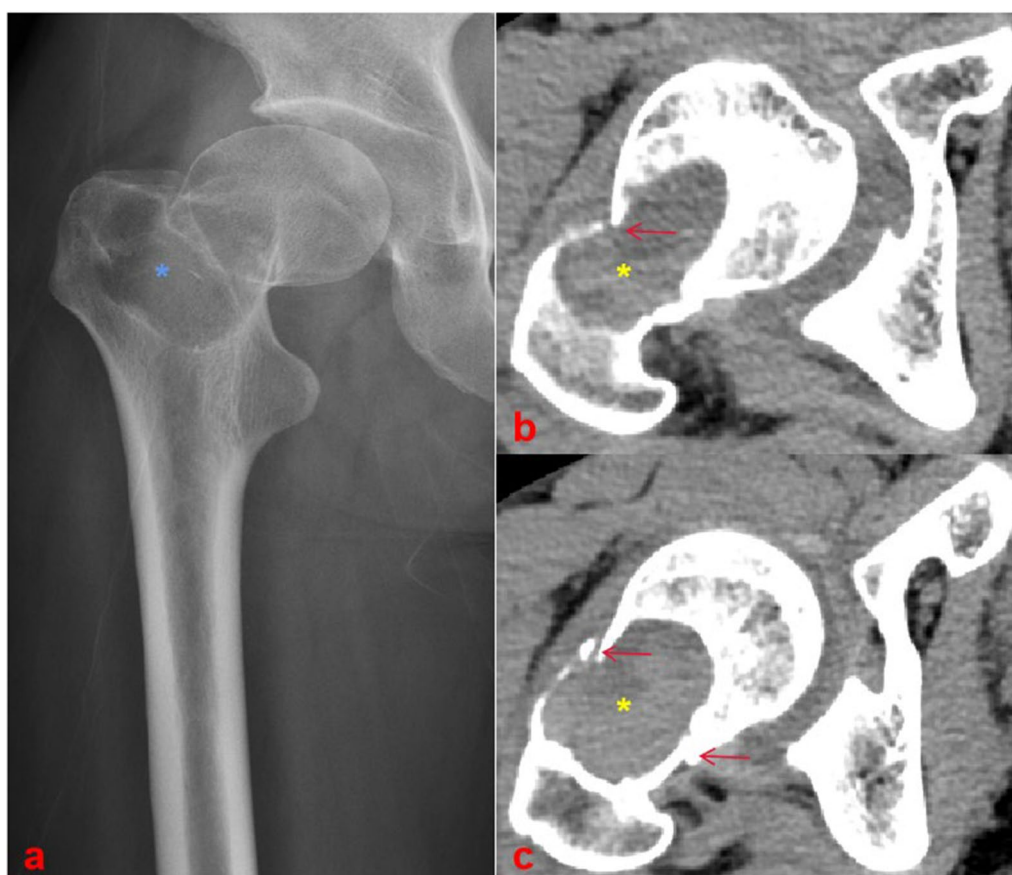


Fig. 6 The radiographic images of the neck of the right femur in Patient 3. **A** Preoperative anteroposterior X-ray showed a typical ground-glass appearance (blue asterisk) and pathological bone fractures of the neck of the right femur. **B** and **C** Preoperative computed tomography image showing pathological bone fractures (red arrows) and cystic lesion of the site (yellow asterisks)



Fig. 7 Fibrous dysplasia accompanied with bone cyst in Patient 4. **A** Preoperative radiographic image showing a typical ground-glass appearance of the proximal femur bone (black asterisk). **B** and **C** Axial computed tomography showed a typical proximal femur lesion with a cyst in the right proximal femur and femoral neck (black asterisks). **D** and **E** Preoperative magnetic resonance imaging. Coronal and sagittal T2-weighted images showed the right proximal femur, and a femoral one showed a proximal femoral lesion with a dominant cystic cavity without any adjacent soft-tissue edema (black asterisks)

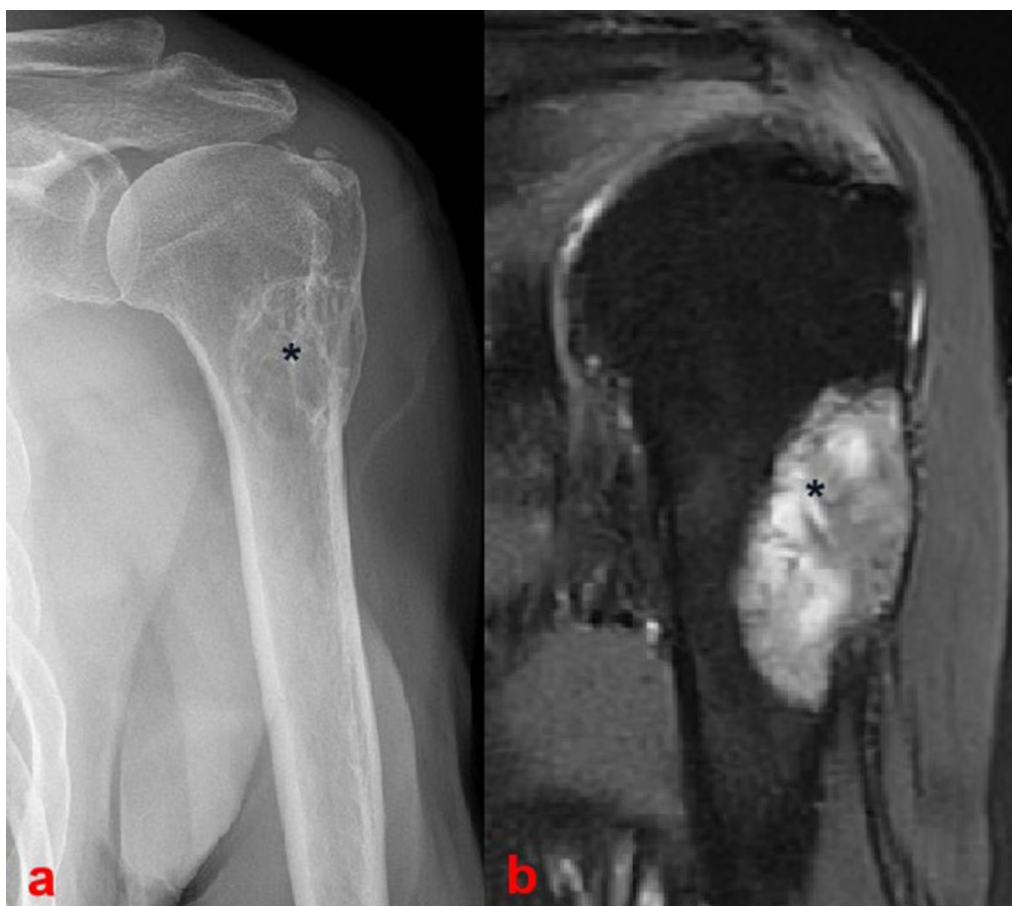


Fig. 8 The radiographic images of fibrous dysplasia in Patient 5. **A** Preoperative anteroposterior X-ray showed a typical ground-glass appearance (black asterisk). **B** Preoperative coronal magnetic resonance imaging. The preoperative sagittal T2-weighted image showed multiple high intensities in the medullary space of humeri bone and a cystic lesion of the site (black asterisk)

fibrous dysplasia is hard to determine, although previous data estimate the incidence rate of fibrous dysplasia worldwide to be 5000–10,000 cases [8]. Additionally, the published literature suggested that there is not a significant difference between the two sexes for those affected by fibrous dysplasia. It is well accepted that this clinical entity compromises 5–7% of all primary bone tumors [9]. In fact, relatively early literature reported that the peak incidence of fibrous dysplasia is often in adolescents and young adults, and it has age-related self-limiting characteristics attributing to the number of mutant cells, often decreasing as age increases [6, 7, 10].

Monostotic fibrous dysplasia may affect simply every bone in the skeleton, tubular bone, flat bone, and the craniofacial bone. In fact, craniofacial sites are more frequently involved in approximately 30% of cases of fibrous dysplasia, and the sites including the vertebrae, radius, fibula, short tubular bones of the hand and foot, and pelvic bones are rarely affected [11]. Our cases of fibrous dysplasia involved the femur (5/7), fibula (1/7), and

humeri (1/7). Notably, the femur was the most frequent site of lesion, followed by the tibia, which was supported by a multi-center clinicopathological study by the European Pediatric Orthopedic Society [8, 11]. There were not cases with McCuneAlbright syndrome or Mazabraud syndrome in our reported cases. In a prior report from the Mayo Clinic *et al.*, the jaw and the long bones were mainly affected in female patients, whereas the craniofacial bones and costae were mainly involved in male patients [12]. In our series, two male patients had mainly long tubular bone involvement (Patients 6 and 7).

Histological analyses of fibrous dysplasia lesions suggested spongy *de novo* bone formation, and unmineralized cancellous bony structures embedded in the marrow stroma [13]. Thus, these trabeculae formed by the above marrow stroma were not surrounded by osteoblasts, causing the appearance of immature bone. The microscopic presenting characteristics of the tumor tissue of fibrous dysplasia includes narrow, circular, irregular, immature bone trabeculae, often shaped like



Fig. 9 The radiographic images of fibrous dysplasia in Patient 6. **A** Preoperative anteroposterior X-ray showed a typical ground-glass appearance (black asterisk). **B** Preoperative coronal magnetic resonance imaging. The preoperative sagittal T2-weighted image showed high intensities in the cystic lesion site of the femur bone (black asterisk)

a fish hook, which were scattered throughout fibrous tissue [14]. This net-like bone showed a paucity of bony trabeculae interspersed within the fibrotic marrow spaces, which gave it the appearance of Chinese characters or an alphabet soup of thin and curvy lines [15]. There are differences from case to case and from area to area in regard to the number, distribution, and maturity of bone trabeculae of fibrous dysplasia. A study found osteoblasts may be present surrounding the reactive bone trabeculae in cases with pathological bone fracture or recurrence following initial interventions, as well [8]. One case of fibrous dysplasia in our patients presented with pathological bone fracture without reactive bone proliferation in a CT image. A secondary bone cyst mimicking cystic degeneration, hemorrhage, and foamy histiocytes were also found in our cases [6]. Some research showed that sometimes intral-lesional hemorrhage may affect osteoclasts, ultimately

leading to diffusion of myxoid degeneration in the fibrous stroma [6, 16].

Although fibrous dysplasia is considered to be a benign disease, malignant transformation is reported in 0.4–6.7% of these cases in the published literature [17, 18]. Such malignant transformation may cause osteosarcoma, chondrosarcoma, and fibrosarcoma formation, yet it is more frequent in the polyostotic form [19]. In addition, some cases may present with recurrent fibrous dysplasia. In our series, one case presented with recurrent monostotic fibular fibrous dysplasia during follow-up, and the patient then agreed to undergo radical excision with reconstruction of fibularis longus allograft combined with fibular locking plate and screws.

Regarding radiological characteristics of fibrous dysplasia, these images differ from X-ray, CT, MRI, single-photon emission computed tomography (SPECT), and positron emission tomography-computed tomography

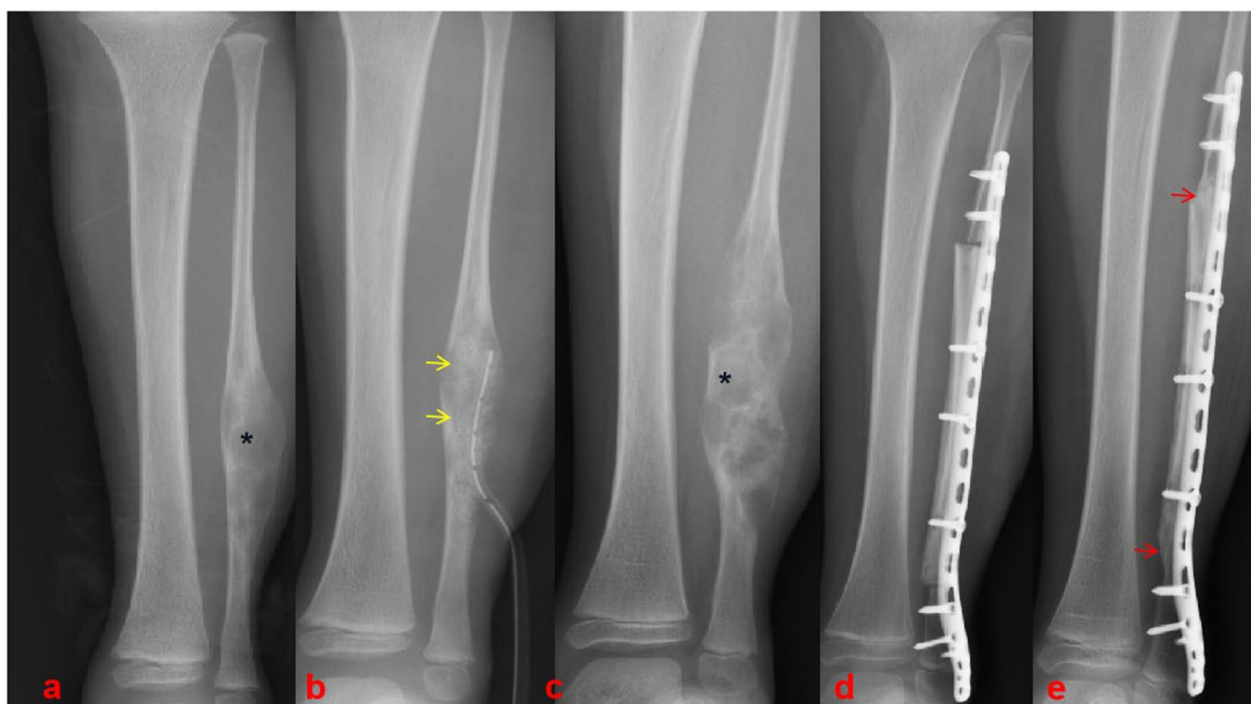


Fig. 10 A therapeutic strategy of recurrent fibular fibrous dysplasia. **A** The first preoperative anteroposterior X-ray showed a ground-glass appearance and partial radiolucent lesions with clear borders around soft tissue (black asterisk). **B** The first postoperative anteroposterior X-ray showed a radiographically apparent bone graft refilling of ground-glass and partial radiolucent sites (yellow arrows) in X-ray images compared with preoperative X-ray images, as shown in Fig. 10 A. **C** The postoperative anteroposterior X-ray showed recurrent fibular fibrous dysplasia on follow-up (black asterisk). **D** Radical resection of the lesion and a fibularis longus bone allograft combined with fibular locking plate and screws were used for reconstruction of the left fibula. **E** A strong autogenous fibular bone fused to allogenic fibular bone was observed (red arrows), and the internal fixation was removed by a surgeon

(PET-CT). Conventional radiography of fibrous dysplasia often presents one or more general radiologic features, including ground-glass appearance, cystic or sclerotic lesions, or mixed cystic and sclerotic lesions [20]. The well-circumscribed margins of fibrous dysplasia may or may not have a sclerotic border, whereas the expanded lesions are accompanied with a thick thin shell showing small perforations with or without endosteal scalloping [21]. These radiological features of fibrous dysplasia also showed up in our cases. However, a soft tissue mass is not a radiological feature of fibrous dysplasia.

As a useful tool for assessment of lesion regions, CT can detect subtle undisplaced fractures, even if this is associated with the complex anatomy of the skeletal structure, craniofacial lesions, pelvis, and spine. An expansile nature, lytic lesions, sclerotic rim formation, and contour deformity with bone remodeling are also shown in CT images. Malignant transformation should be considered if the presence of an extra-osseous soft tissue mass with bony destruction was detected using CT [22].

Fibrous dysplasia has non-specific features on MRI. Thus, MRI allows differentiation of fibrous dysplasia from

a cyst lesion. Research has revealed that, on T1-weighted images, the signal intensities of fibrous dysplasia were hypointense and/or intermediate, whereas some hyperintense signal intensities were also observed in these weighted images. On T2-weighted images, the signal intensities of fibrous dysplasia were extremely variable, such that the proportion of signal intensity was distributed almost evenly in all sequences [22, 23]. In addition, lesions in fibrous dysplasia showed at least some enhancement on contrast-enhanced images, as proved by Kinnunen AR *et al.* [23]. Abnormal imaging findings are also shown on SPECT and/or PET-CT, which demonstrate increased tracer uptake at the foci of skeletal disease [24].

Fibrous dysplasia lesions are almost invariably benign with very rare malignant transformation, whereas recurrence can occur owing to inappropriate therapeutic methods. Even so, some studies have suggested that fibrous dysplasia has age-related self-limiting characteristics and rare malignancy of transformation, attributed to the number of mutant cells often decreasing with age [6, 8, 25]. Thus, the treatment of fibrous dysplasia is also subject to debate. The initial approach is typically

Table 1 Clinicopathological findings of cases ($n = 7$)

Variable	Types	No. of cases
Gender	Female	4
	Male	3
Age	≤ 18 years	3
	18–40 years	2
	≥ 40 years	2
Type	Monostotic	7
Tumor site	Femur	5
	Humerus	1
	Fibula	1
Histological variant	Classical fibrous dysplasia	4
	Fibrous dysplasia and bone cyst	1
	Fibrous dysplasia and aneurysmal-bone-cyst-like changes	2
Secondary aneurysmal bone cyst	Yes	2
	No	5
Pathological fracture	Yes	1
	No	6
Surgery	Curettage	4
	Resection	2
Recurrence	Yes	1
	No	6
Malignant transformation	Yes	0
	No	7

Table 2 Clinical data of recurrent cases

Case	Age (years)	Sex	Site	First surgical procedure	Recurrences	Time to first recurrence	Management of recurrence	Follow-up (months)
1	49	F	Femur	Curettage combined with allogenic bone grafting	No	—	—	15
2	37	M	Femur	Resection and total hip replacement	No	—	—	1
3	35	F	Femur	Curettage combined with allogenic bone grafting and internal fixation	No	—	—	12
4	17	M	Femur	Curettage combined with allogenic bone grafting	No	—	—	3
5	71	F	Humerus	Curettage combined with allogenic bone grafting and internal fixation	No	—	—	12
6	16	F	Femur	Curettage combined with allogenic bone grafting and internal fixation	No	—	—	15
7	8	M	Fibula on the site column	Curettage combined with allogenic bone grafting and internal fixation	Yes	2 years	1. Resection and fibularis longus allograft transplantation combined with internal fixation 2. Removal of internal fixation	39

F, female; M, male

conservative therapy aiming to avoid physical disability. As a proposed treatment, therapeutic drugs including calcitonin, bisphosphonates, and denosumab have been used in some patients to improve analytical parameters and alleviate pain [26–28]. However, no long-term evidence backs the use of the abovementioned drugs for control of the disease. The most common indications are for cosmetic reasons, especially in the craniomaxillofacial type of fibrous dysplasia, due to neurovascular compression or orbital involvement. Some authors suggested that surgical curettage with or without bone grafting is the method of treatment in a majority of cases of fibrous dysplasia of long bone; nevertheless, radical excision may be more suitable when fibrous dysplasia recurs after conservative surgery and medical treatment [6, 7, 29]. In our case series, most of patients accepted curettage associated with bone grafting with or without internal fixation. One patient presenting with recurrent fibular fibrous dysplasia underwent radical excision associated with fibularis longus allograft transplantation combined with fibular locking plate.

Conclusion

A rare benign skeletal disorder caused by interruption of regular bone growth, fibrous dysplasia is usually benign in nature. Thus, appropriate diagnosis and personalized treatment planning may prevent recurrence and malignant transformation.

Acknowledgements

None.

Author contributions

Xie LL designed, drafted, and revised the manuscript and approved the final version. Liu MH, Zhu HX, and Fu Lei acquired data and played an important role in interpreting the results. All authors contributed equally to this work and have read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

The study protocol was approved by the institutional review board of Hunan University of Medicine General Hospital, and all medical activities were performed in accordance with approved guidelines and regulations.

Consent for publication

Written informed consent was obtained from the patient and/from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors have declared that no competing interest exists.

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References

- Kim HY, Shim JH, Heo CY. A rare skeletal disorder, fibrous dysplasia: a review of its pathogenesis and therapeutic prospects. *Int J Mol Sci*. 2023;24(21):15591. <https://doi.org/10.3390/ijms242115591>.
- Riminucci M, Robey PG, Saggio I, Bianco P. Skeletal progenitors and the GNAS gene: fibrous dysplasia of bone read through stem cells. *J Mol Endocrinol*. 2010;45(6):355–64. <https://doi.org/10.1677/JME-10-0097>.
- Sauhta R, Makkar D. A closer look at fibrous dysplasia femur fracture five-year follow-up: a unique case. *J Orthop Case Rep*. 2024;14(2):155–9. <https://doi.org/10.13107/jocr.2024.v14.i02.4254>.
- Zhao X, Deng P, Iglesias-Bartolome R, Amornphimoltham P, Steffen DJ, Jin Y, Molinolo AA, de Castro LF, Ovejero D, Yuan Q, Chen Q, Han X, Bai D, Taylor SS, Yang Y, Collins MT, Gutkind JS. Expression of an active Gαs mutant in skeletal stem cells is sufficient and necessary for fibrous dysplasia initiation and maintenance. *Proc Natl Acad Sci U S A*. 2018;115(3):E428–37. <https://doi.org/10.1073/pnas.1713710115>.
- Jayasoorya A, Pisulkar G, Samal N, Taywade S, Vasavada SN. A rare case of monostotic fibrous dysplasia of the femoral neck with pathological fracture: a case report. *Cureus*. 2023;15(11): e49085. <https://doi.org/10.7759/cureus.49085>.
- Xie LL, Yuan X, Zhu HX, Pu D. Surgery for fibrous dysplasia associated with aneurysmal-bone-cyst-like changes in right proximal femur: a case report. *World J Clin Cases*. 2023;11(26):6170–5. <https://doi.org/10.12998/wjcc.v11.i26.6170>.
- Xie LL, Yuan X, Zhu HX, Fu L, Pu D. Fibula allograft transplantation combined with locking plate for treatment of recurrent monostotic fibular fibrous dysplasia: a case report. *World J Clin Cases*. 2023;11(33):8050–7. <https://doi.org/10.12998/wjcc.v11.i33.8050>.
- Lail RA, Majeed A. Clinical presentation and outcome of fibrous dysplasia in patients attending Sahiwal Teaching Hospital, Punjab. *J Univ Coll Med Dent*. 2022;1:20–3.
- Chapurlat RD, Meunier PJ. Fibrous dysplasia of bone. *Baillieres Best Pract Res Clin Rheumatol*. 2000;14(2):385–98. <https://doi.org/10.1053/berh.1999.0071>.
- Ozek C, Gundogan H, Bilkay U, Tokat C, Gurler T, Songur E. Craniomaxillofacial fibrous dysplasia. *J Craniofac Surg*. 2002;13(3):382–9. <https://doi.org/10.1097/00001665-200205000-00004>.
- Cohen MM Jr. Fibrous dysplasia is a neoplasm. *Am J Med Genet*. 2001;98(4):290–3. [https://doi.org/10.1002/1096-8628\(20010201\)98:4%3c290::aid-ajmg1112%3e3.0.co;2-f](https://doi.org/10.1002/1096-8628(20010201)98:4%3c290::aid-ajmg1112%3e3.0.co;2-f).
- Unni KK, Inwards CY. Dahlin's bone tumors: general aspects and data on 11087 cases. 6th ed. Philadelphia: Lippincott Williams and Williams; 2010. p. 310–6.
- Riminucci M, Liu B, Corsi A, Shenker A, Spiegel AM, Robey PG, Bianco P. The histopathology of fibrous dysplasia of bone in patients with activating mutations of the G_s α gene: site-specific patterns and recurrent histological hallmarks. *J Pathol*. 1999;187:249–58.
- Ko JH, Park GJ, Lee KB. Multiple calcaneal fibrous dysplasia: a case report. *Medicine (Baltimore)*. 2019;98(51): e18389. <https://doi.org/10.1097/MD.00000000000018389>.
- Torrico-Acha X, Llaguno-Rubio JM. New technologies in oral radiology as a diagnostic aid for monostotic fibrous dysplasia: a review. *Rev Cient Odontol (Lima)*. 2021;9(4): e089. <https://doi.org/10.21142/2523-2754-0904-2021-089>.
- Bhattacharya S, Mishra RK. Fibrous dysplasia and cherubism. *Indian J Plast Surg*. 2015;48(3):236–48. <https://doi.org/10.4103/0970-0358.173101>.
- Doganavsargil B, Argin M, Kececi B, Sezak M, Sanli UA, Oztup F. Secondary osteosarcoma arising in fibrous dysplasia, case report. *Arch Orthop Trauma Surg*. 2009;129:439–44.
- Ruggieri P, Sim FH, Bond JR, Unni KK. Malignancies in fibrous dysplasia. *Cancer*. 1994;73:1411–24.

19. Qu N, Yao W, Cui X, Zhang H. Malignant transformation in monostotic fibrous dysplasia: clinical features, imaging features, outcomes in 10 patients, and review. *Medicine (Baltimore)*. 2015;94: e369.
20. Liu XX, Xin X, Yan YH, Ma XW. Imaging characteristics of a rare case of monostotic fibrous dysplasia of the sacrum: a case report. *World J Clin Cases*. 2021;9(5):1111–8. <https://doi.org/10.12998/wjcc.v9i5.1111>.
21. Zhang Y, Zhang C, Wang S, Wang H, Zhu Y, Hao D. Computed tomography and magnetic resonance imaging manifestations of spinal monostotic fibrous dysplasia. *J Clin Imag Sci*. 2018;8:23. https://doi.org/10.4103/jcis.JCIS_20_18.
22. Park SK, Lee IS, Choi JY, Cho KH, Suh KJ, Lee JW, Song JW. CT and MRI of fibrous dysplasia of the spine. *Br J Radiol*. 2012;85(1015):996–1001. <https://doi.org/10.1259/bjr/81329736>.
23. Kinnunen AR, Sironen R, Sipola P. Magnetic resonance imaging characteristics in patients with histopathologically proven fibrous dysplasia—a systematic review. *Skeletal Radiol*. 2020;49(6):837–45. <https://doi.org/10.1007/s00256-020-03388-x>.
24. Jreige M, Hall N, Becce F, Aubry-Rozier B, Gonzalez Rodriguez E, Schaefer N, Prior JO, Nicod LM. A novel approach for fibrous dysplasia assessment using combined planar and quantitative SPECT/CT analysis of Tc-99m-diphosphonate bone scan in correlation with biological bone turnover markers of disease activity. *Front Med (Lausanne)*. 2022;9:1050854. <https://doi.org/10.3389/fmed.2022.1050854>. (Erratum.In:FrontMed(Lausanne).2023Mar,07(10),pp.1171916).
25. Özşen M, Yalçinkaya Ü, Bilgen MS, Yazıcı Z. Fibrous dysplasia: clinic pathologic presentation of 36 cases. *Türk Patoloji Derg*. 2018;34(3):234–41. <https://doi.org/10.5146/tjpath.2018.01428>.
26. Szymczuk V, Taylor J, Boyce AM. Craniofacial fibrous dysplasia: clinical and therapeutic implications. *Curr Osteoporos Rep*. 2023;21(2):147–53. <https://doi.org/10.1007/s11914-023-00779-6>.
27. DiCaprio MR, Enneking WF. Fibrous dysplasia. Pathophysiology, evaluation, and treatment. *J Bone Joint Surg Am*. 2005;87(8):1848–64. <https://doi.org/10.2106/JBJS.D.02942>.
28. Florez H, Peris P, Guañabens N. Fibrous dysplasia. Clinical review and therapeutic management. *Med Clin (Barc)*. 2016;147(12):547–53. <https://doi.org/10.1016/j.medcli.2016.07.030>.
29. Baiomy AABA, Elsayed SA, Abdelfattah MA, Khalifa FA. Management of massive mandibular fibrous dysplasia with radical excision and different immediate reconstructive modalities: case series report. *J Maxillofac Oral Surg*. 2022;21(4):1311–9. <https://doi.org/10.1007/s12663-021-01660-8>.

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